# Ruptured Isolated Spinal Artery Aneurysms

## Report of Two Cases and Review of the Literature

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**Key words:** arterial dissection, spinal artery, spinal aneurysm, spinal subarachnoid haemorrhage

**Summary**

*Isolated spinal artery aneurysms are exceedingly rare vascular lesions thought to be related to dissection of the arterial wall. We describe two cases presenting with spinal subarachnoid haemorrhage that underwent conservative management.*

*In the first patient the radiculomedullary branch involved was feeding the anterior spinal artery at the level of D3 and thus, neither endovascular nor surgical approach was employed. Control angiography was performed at seven days and at three months, demonstrating complete resolution of the lesion. In our second case, neither the anterior spinal artery or the artery of Adamkiewicz could be identified during angiography, thus endovascular management was deemed contraindicated. Magnetic resonance imaging showed a stable lesion in the second patient. No rebleeding or other complications were seen.*

*In comparison to intracranial aneurysms, spinal artery aneurysms tend to display a fusiform appearance and lack a clear neck in relation to the likely dissecting nature of the lesions.*

*Due to the small number of cases reported, the natural history of these lesions is not well known making it difficult to establish the optimal treatment approach. Various management strategies may be supported, including surgical and endovascular treatment, but It would seem that a wait and see approach is also viable, with control angiogram and treatment decisions based on the evolution of the lesion.*

## Introduction

Aneurysms arising from spinal arteries are extremely rare and are typically related to lesions that induce an increase in blood flow through the arteries, such as spinal cord arteriovenous malformations (ScAVMs), arteriovenous fistulas, coarctation of the aorta [1], bilateral vertebral artery occlusion or Moyamoya disease. When a spinal artery aneurysm is not related to any of these conditions, then it is referred as an isolated spinal aneurysm [2].

Rupture of an isolated spinal aneurysm is a rare cause of subarachnoid haemorrhage (SAH) accounting for less than 1% of all cases reported in the literature [3]. The most frequent aetiologies of spinal SAH include rupture from ScAVMs followed by intraspinal tumours. We describe two cases of spinal SAH caused by rupture of an isolated spinal artery aneurysm.

## Case Report

### *Patient 1*

A 37-year-old woman without a relevant medical history presented with a sudden onset of thoracic pain, followed by cervical pain and headache. On clinical examination neck stiffness was found without neurological impairment. A non-enhanced head computed tomography (CT) scan showed SAH involving primarily the infratentorial space (Figure 1). With the suspicion of spinal bleeding a magnetic

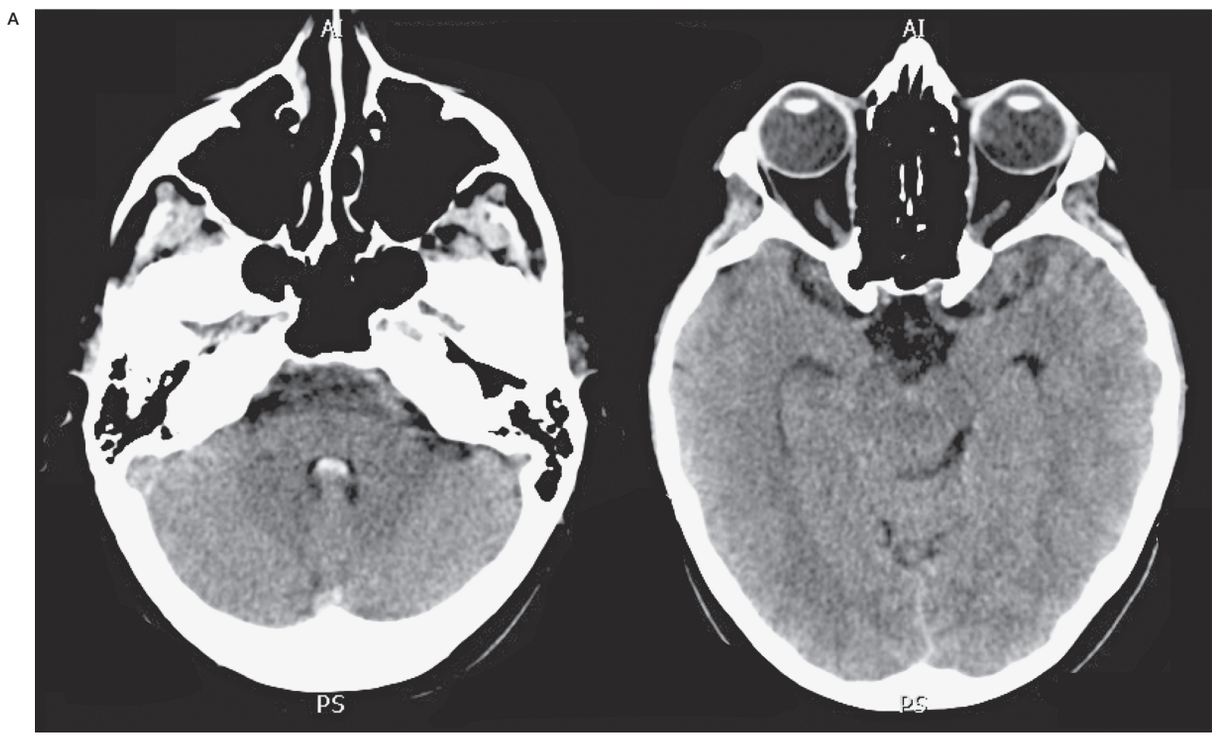


*Figure 1* Initial head CT scan. Axial slices at the level of the fourth ventricle (A) and mescencephalus (B). Hyperdense (70HU) occupation of the forth ventricle and interpeduncular cistern is seen, corresponding to blood products in the subarachnoid space. No other anomalies were seen in the remaining study, including and intracranial CTA (not shown).

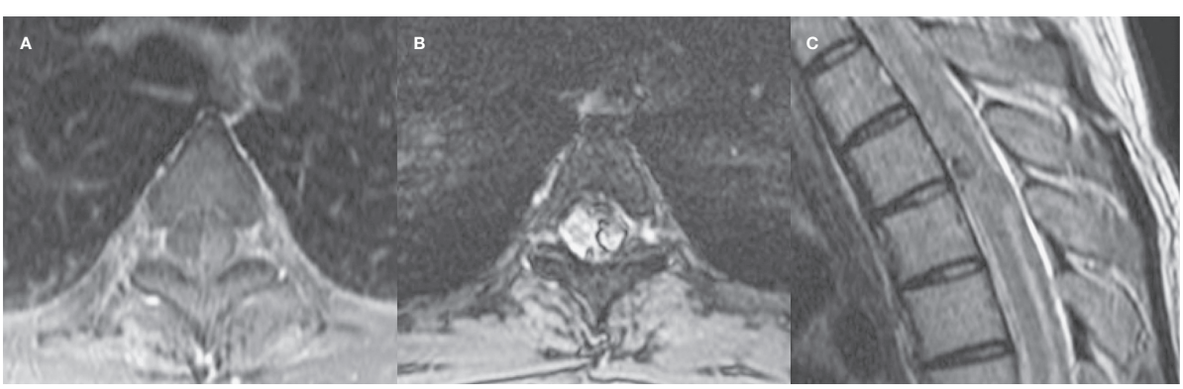


*Figure 2* Axial fat-suppressed T1W (A), T2* (B) and sagittal T2W (C), MR of the spine demonstrating a decreased T2 signal around the spinal cord in relation to SAH, as well as an 8mm nodular lesion at the level of T3, located in the left lateral aspect of the spinal cord in an extramedullary situation. This lesion presents high SI in the T1 fat-suppressed sequence along with magnetic susceptibility artifacts in the T2* sequence. No signal abnormality was noted in the spinal cord.

resonance imaging (MRI) study was acquired, revealing intradural bleeding along with an 8 mm low signal intensity (SI) nodular lesion at the level of D3 (Figure 2). No other anomalies were noted. Spinal angiography was performed the following day to look for a vascular pathology. Selective injection of the left superior intercostal artery revealed a fusiform aneurysm in the intradural segment of a radiculomedullary artery at the level of D3 feeding the anterior spinal artery (Figure 3). No other vascular anomalies were seen. A diagnosis of dissecting aneurysm was established. Conservative management was proposed with an uneventful clinical course.

Control angiography performed at seven days demonstrated a complete resolution of the lesion. The patient was subsequently discharged at day 14. Angiographic control at three months was normal.

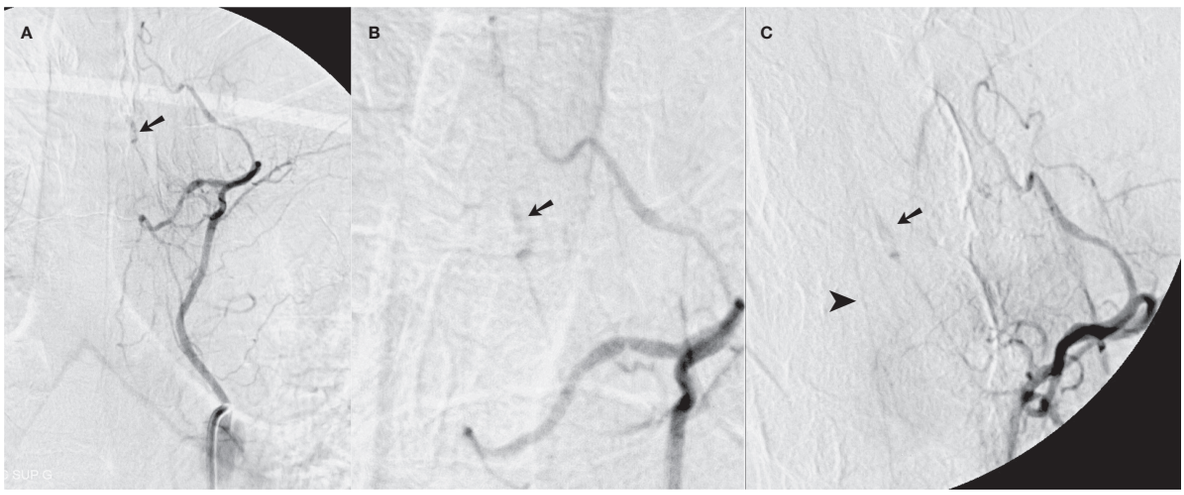


*Figure 3* Digital subtraction angiography, frontal (A,B) and oblique (C) views. Selective catheterism of the left supreme intercostal artery feeding the T3 and T4 intercostal arteries. A radiculomedullary branch arising from T4 shows a focal fusiform dilatation compatible with a false aneurysm (arrow in A-C). The radicullomedulary artery involved is seen feeding the anterior spinal artery (arrowhead in C). No other vascular anomalies were seen. The Adamkiewicz artery was fed by T9 (not shown). In the absence of other vascular anomalies and given the characteristic imaging findings, an isolated spinal artery aneurysm must be considered.

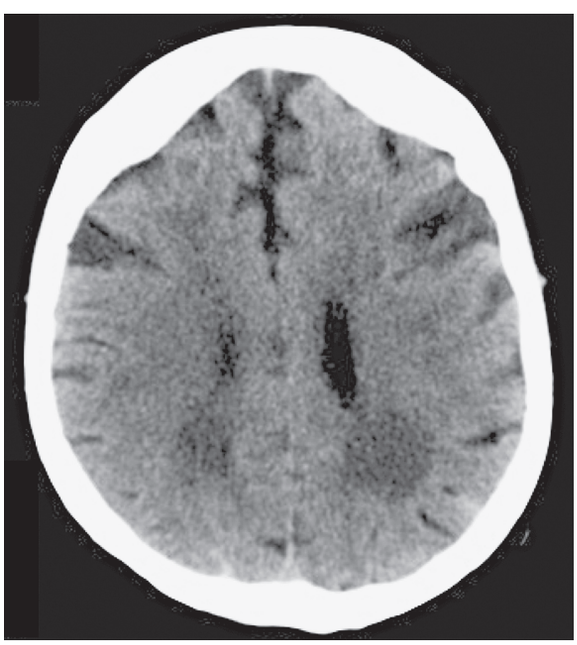

*Figure 4* Non-enhanced head CT scan. Axial slice at the level of the corona radiata show hyperdense occupation of cortical parietal sulci, corresponding to SAH. No other anomalies were seen in the remaining study, including and intracranial CTA (not shown).

*Patient 2*

A 72-year-old woman with a medical history of arterial hypertension, diabetes mellitus, chronic renal failure and four lumbar discectomies experienced a sudden headache associated with nausea and vomiting on the previous day. She was admitted to a nearby hospital where physical examination revealed neck stiffness. Neurological examination was unremarkable. Non-enhanced head CT was normal. On subsequent interrogation the patient also referred cervical and dorsal pain and so a spinal MRI was acquired but due to movement artifacts it was not of diagnostic quality. Cerebrospinal fluid study was positive for SAH. The patient was then transferred to our institution where a new head CT revealed SAH involving the cortical parietal sulci (Figure 4). Intracranial CT angiography did not reveal any abnormality. Spinal

MRI was repeated showing occupation of the spinal canal, corresponding either to a subarachnoid haematoma or subdural haematoma associated with a 5 mm low SI nodular lesion at the level of D10 (Figure 5). Spinal angiography was performed the following day to look for a vascular pathology. Selective injection of the left D11 intercostal artery revealed a fusiform aneurysm in the intradural segment of a radiculopial artery (Figure 6). No other vascular anomalies were seen. The angiographic finding was interpreted as a dissecting aneurysm. Conservative management was proposed. During the course of admission the patient presented an increase in cervical pain, delirium and fluctuating lower limb strength. Control MRI studies showed a minimal decrease in the size of the nodular lesion corresponding to the aneurysm along with the expected degradation of blood products, without evidence of rebleeding or increase in intrathecal haematoma size. Finally there was a slow recovery of her mental status and normalization of the neurological examination. The patient was discharged at week 3. A control angiogram is pending.

## Discussion

Spinal artery aneurysms are heterogeneous lesions with variable pathophysiological characteristics, morphological features, and anatomical locations [4]. They represent a rare entity and are typically related to lesions that induce an increase in blood flow through the arteries.

Isolated spinal artery aneurysms are exceedingly rare, and are thought to be related to dissection of the arterial wall probably associated with underlying states leading to inflammation of the vascular wall [2], as reported in at least five previous cases (Pseudoxanthoma elasticum [5], fibromuscular dysplasia [6], Behcet's disease [7], systemic candidiasis and rheumatoid arthritis [8]), interestingly all of them located in the anterior spinal artery.

The first case of an isolated spinal aneurysm was described by Babonneix and Wediez in 1930 [9] and to our knowledge only 41 patients with this entity have been reported in the literature. In 23 of these cases the aneurysm was located in the anterior vascular spinal axis, 11 in the posterior spinal artery, five at a radicular or radiculopial artery and one in a lateral spinal artery. In two cases the exact location was not specified [2].

The different pathophysiology of spinal artery aneurysms gives them differentiating characteristics from intracranial lesions. They tend to occur along the course of small arteries, less affected by atherosclerosis and with no relation to the branching points. A fusiform appearance and lack of a clear neck are also common features, in relation to the likely dissecting nature of the lesions [10].

Due to the small number of cases reported, the natural history of these lesions is not well-known. Spinal aneurysms typically present with haemorrhage, as in the two patients described here. After haemorrhage, compression with symptoms related to the level of the lesion is a common manifestation [11]. Thoracolumbar haemorrhage may result in back pain, motor weakness and sensory loss of the lower extremities. Bleeding at the cervical level may present with overlapping symptoms from those caused by intracranial SAH.

### *Diagnosis: CT and MRI*

Isolated spinal artery aneurysms should be considered when there are spinal symptoms with subarachnoid haemorrhage and no other source of bleeding is identified, and or when the bleed is limited to or largely predominant in the spine. A complete spinal MRI including dynamic contrast-enhanced MR angiography and post contrast sequences should be performed and scrutinized for low SI T2 nodular lesions representing an aneurysm as in our cases. Enhancing areas along the surface of the spinal cord representing contrast stagnation inside the aneurysm [2] may be seen after contrast administration.

CT angiography may also be performed to better visualize the relation between the aneurysm and the spinal canal.

Angiography constitutes the most sensitive examination for the diagnosis of isolated spinal aneurysms, not only allowing the visualization of arterial feeders, but also permitting the exclusion of associated pathologies. Faced with the complexity and variety of spinal aneurysms, it is extremely important to precisely locate the aneurysm with selective injections to understand its exact relation to the main spinal arterial axis [4]. Vascular supply of the spinal cord may be summarized as follows: in the dorsal spine, each segmental artery bifurcates into a dorsal branch and an intercostal branch. The latter supplies the rib, adjacent muscle and other soft tissues.

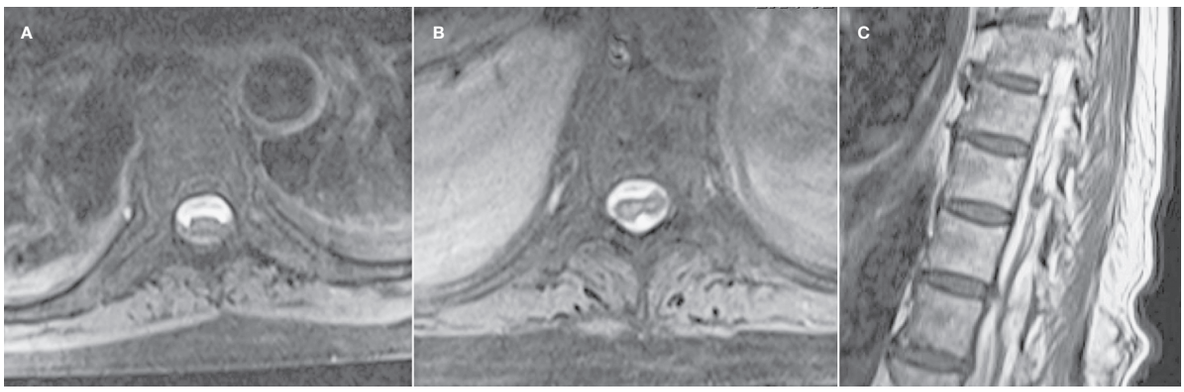


*Figure 5* Axial fat-suppressed T1W (A and B) and sagittal T1W (C) MR of the spine demonstrating a high SI T1 occupation of the spinal canal corresponding to blood products, either a subarachnoid haematoma or subdural haematoma. A 5 mm low SI nodular lesion is seen at the level of T10, located in the left lateral aspect of the spinal cord, in an extramedullary location. No signal abnormality was noted in the spinal cord.

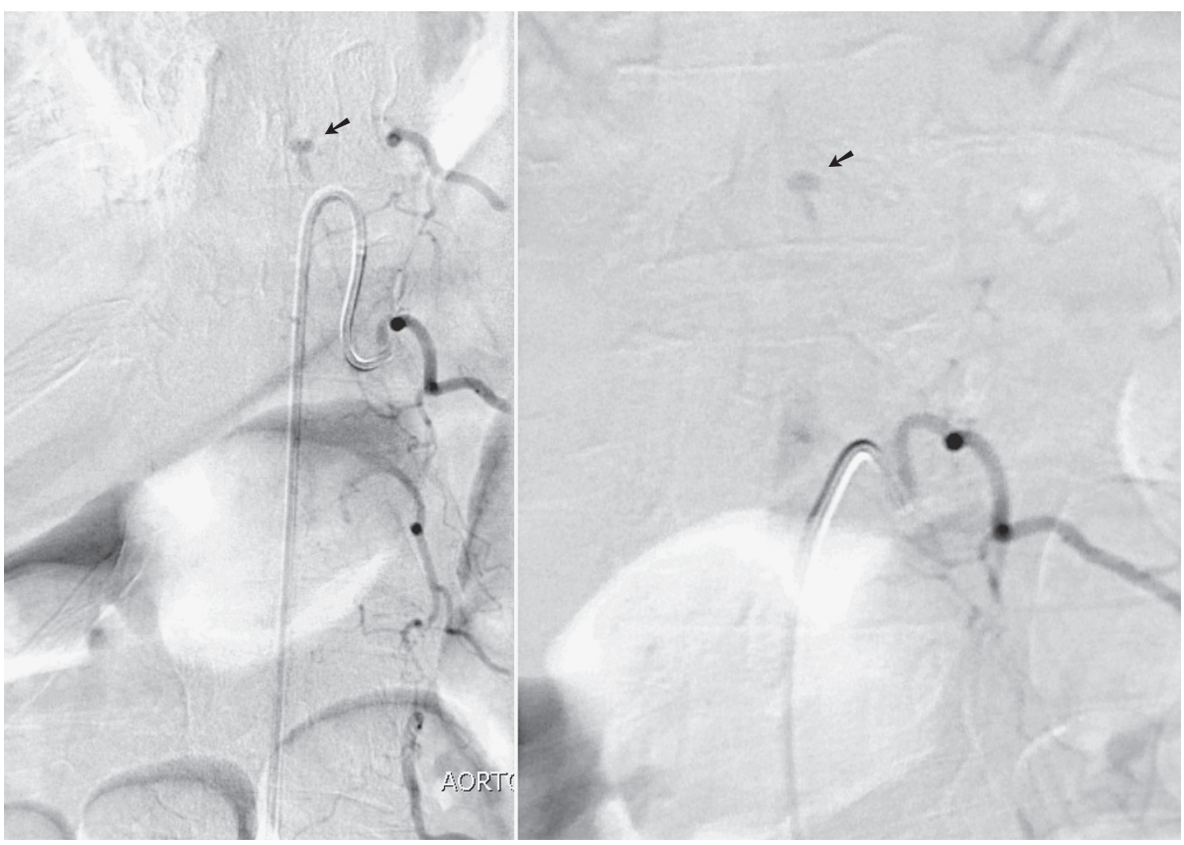


*Figure 6* Digital subtraction angiography, frontal view (A) and frontal magnified view (B). Selective catheterism of the left D11 intercostal artery. A focal fusiform dilation of a radiculopial artery located at the level of D10 is seen (arrow in A and B). The lesion is compatible with a false aneurysm. Neither the anterior spinal nor the Adamkiewicz arteries were seen. No other vascular anomalies were identified.

The dorsal branch via the neural foramen sends branches to supply the local epidural and dural elements, as well as a radicular artery to nourish the nerve root [12]. This basic system varies in the cervical, upper thoracic and sacral segments. In total, four to eight of these segmental arteries gain access to the anterior spinal artery and ten to 20 supply the posterior spinal arteries effectively turning into radiculomedullary arteries because they also supply a large segment of the

spinal cord. The two most significant radiculomedullary arteries are the artery of cervical enlargement and the artery of lumbar enlargement, also known as the artery of Adamkiewicz. Thus, isolated spinal artery aneurysms may be located in the radicular artery, radiculopial artery, radiculomedullary artery or directly over the anterior or posterior spinal arteries. As previously stated, most of the lesions compromise the anterior vascular axis, and are located at the thoracic region.

*Management*

Management of isolated spinal artery aneurysms remains controversial, mostly due to the rarity of these lesions. The options include a surgical approach, endovascular embolization, or conservative treatment. The exact localization of the lesion is of prime importance for planning any intervention.

The surgical approach is more commonly employed in posterior localized aneurysms due to their dorsolateral and superficial location [2,10]. Because of the small size of the arteries involved and the dissecting nature of these lesions, preservation of the artery is difficult to achieve, and thus, surgical resection of the affected artery is usually performed [10]. Still, some cases of parent artery preservation have been reported using mucin wrapping or surgical vessel reconstruction [13].

Endovascular treatment with embolization using coils or liquid embolic agents is possible but as with the surgical approach poses the same difficulties regarding the preservation of the parent artery, and furthermore, it may be hindered by the small size of the radiculopial arteries. If endovascular treatment is to be considered, it should be remembered that the specific morphologic features of spinal artery aneurysms usually preclude parent vessel preservation; thus, a complete mapping of the vascular supply of the spinal cord must be performed. Careful identification of flow distal to the aneurysm should be assessed before deciding whether to sacrifice the affected artery. If the affected artery presents distal flow supplying the spinal cord, especially the anterior axis, parent vessel sacrifice should be avoided.

Spinal cord infarction constitutes the most fearful complication when dealing with these types of vascular lesions, both in surgical and endovascular treatments.

It could be secondary to obliteration of a dominant radiculomedullary artery or unintended distal embolization.

Conservative medical treatment constitutes another management strategy especially in cases where the parent artery cannot be preserved, as well as in cases where the surgical or endovascular approaches are contraindicated because of the patient's condition. A total of ten cases with conservative management have been reported in the literature. Of these, two patients died [14,15] with the remainder showing stable lesions or complete resolution of the aneurysms [5,16–19].

The two cases we presented underwent conservative management. The first patient was neurologically intact; because the involved radiculomedullary branch was feeding the anterior spinal artery neither endovascular nor surgical approach was employed. Control angiogram at seven days showed complete resolution of the lesion. The patient recovered completely without any clinical deficit and a late angiographic control at three months confirmed the spontaneous cure of the lesion.

In our second case, neither the anterior spinal artery or the artery of Adamkiewicz could be identified during the angiography, thus endovascular management was deemed contraindicated. On the other hand, due to the age and comorbidities of the patient and the still unknown natural history of isolated spinal aneurysms, surgery was not performed either. MRI performed three weeks later showed a minimal decrease in the size of the lesion, without evidence of rebleeding, and no clinical deterioration.

## Conclusions

Until more data are available on the natural history of ruptured isolated spinal artery aneurysms, it is difficult to establish the optimal treatment approach and various management strategies may be supported. It would seem that a wait and see approach is viable, with control angiogram and treatment decisions based on the evolution of the lesion. On the other hand, prompt surgical treatment, especially in lesions located in the posterior aspect of the spinal cord and not involving dominant radiculomedullary arteries, seems to be a valid option. Endovascular treatment appears to have a more restricted field of action, with few cases reporting this approach, probably related to technical difficulties and a high possibility for complications.

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